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# Branchial and thyroglossal cysts and fistulae

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# BRANCHIAL & THYROGLOSSAL CYSTS & FISTULAE

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## INTRODUCTION

This thesis will be confined entirely to branchial and thyroglossal duct cysts and fistulas with the purpose of covering the two separate disease entities in one paper. It is also important to mention that because of the close similarity of these two pathological conditions both from the standpoint of the historical background, particularly the embryological background, pathology, clinical aspects and treatment, it is without comment that these two conditions should be discussed together in one paper.

Many articles on the two subjects have appeared in the literature in the past ten to fifteen years, but because of their incompleteness and because they presented nothing new, they were not listed.

## HISTORICAL BACKGROUND OF BRANCHIAL CYSTS & FISTULAE

Cervical fistulae were first reported by Huncyorwski (27) in 1789. Congenital cervical fistulae were described by Dzondi (16) in 1829. But the studies of fistulas of the neck by these two men and others up to the presentations of von Ascherson (20) in 1832 was in its infancy and in a chaotic state.

Von Ascherson (19) began to investigate such conditions more thoroughly. After compiling his

cases of lateral fistulas with all the known cases of fistulas and cysts at that time, von Ascherson concluded that these fistulas resulted from malformed branchial clefts which failed to close or to unite normally. In view of the work of von Rathke, von Ascerson was cognizant of prominent process that developed out of the second branchial arch in vertebrate embryos and that is the most prominent portion of the branchial apparatus: that is, the "Operculum branchiale of von Rathke" (19). Ascherson stated that the external orifice of fistulas pointed up under the operculum and that the fistula was formed by a failure of the cleft boundaries to fuse or close(19). Succeeding authors introduced the conception that a fistula could develop from any of the four clefts. The fact that prompted this deduction was the variety of positions that the external orifice might and did take, being limited, as they knew, only by the region of the styloid process and sternoclavicular junction (19).

According to von Heusinger (19) the internal orifice of all fistulas was situated at the lateral wall of the pharynx at the base of the tongue. He also believed that the location of the external orifice determined which cleft was at fault, and stated that the fourth cleft was responsible in a case in which the ori-

fice as at the sternoclavicular junction, because by dissection he had found that it coursed along the medial border of the sternocleidomastoid muscle to the upper border of the larynx and then made a sharp turn toward the midline, opening at the root of the tongue. Lateral cervical cysts were placed on an embryological basis as being of branchial origin by Rosen in 1859 (27). Among the noteworthy studies of the embryology of the branchial apparatus about the time of von Ascherson were those made by von Rathke, His and Born (19).

Bland-Sutton and von Cussett devised schemas according to which one could determine from which of the four clefts a fistula developed. His doubted the truth and value of these schemas and maintained that the existence of a fistula opening into the sinus praecervicalis but not into the pharynx is normal in the five weeks' embryo. Later the thymus develops from a portion of the sinus praecervicalis forming a connection between the two(19). If, His stated, the existing fistula does not grow downward in the thymus regions but upward, then a true pathological fistula exists (19). His, in 1889, changed his mind and stated that the thymus developed from the pharyngeal pouch and not from the precervical sinus. This sinus was described by His stating that it was formed by the over lapping of the

hyoid arch in front of the thyroid gland.

Rabl refuted His, saying that the fourth cleft could not be a factor in the formation of fistulae, because it is separated from the sinus precervicalis by a thick mesodermal layer. The fistula could not result from the third arch because it is concerned with the formation of the thymus gland and therefore the fistula would have to be in close relationship with the thymus which Hyndman in 1929 (19) stated was not true. Rabl paid special attention to the origin of the viscera or organs of the neck from the second arch, since this arch is larger than the others. Rabl continued his presentation by stating that the second groove consists of a long irregular passage in which two rows of closely opposed epithelial cells divide the pharynx from the outside. He termed this the branchial canal and believed it to be the basis of the formation of fistulae when the epithelial membrane ruptured and therefore allowed communication of the pharynx with the sinus precervicalis (19).

In 1890, Kostaniecki and Milecki studied the literature in detail paying special attention to Rabl's work. They did not agree with the schemas of Heusinger, Bland-Sutton and others that the external opening of the fistula may at one time be high up near the jaw,

then may close and open later in a lower position, and they therefore thought it foolish to say that the position of this opening had such significance as was claimed. The inner opening into the pharynx, however, is constant for any particular cleft or groove as Hyndman pointed out (19). The most frequent and constant internal opening is located in the supra-tonsillar fossa, and therefore Kostaniecki and Milecki concluded from a search of the literature that this corresponded with the second arch, thus agreeing with Rabl (27). They also contended that no branchial cleft could persist in toto, nor could it itself be the cause of a fistula. In this regard Hyndman pointed out that in the case of a complete fistula, the limiting membrane must be ruptured and in this way the pharyngeal pouch must first break into the cervical sinus. He concluded therefore that the complete fistula originates from a rupture of the second pharyngeal pouch into the cervical sinus. Although later work was based on the observations of Kostaniecki and Milecki, some men in the early nineteenth century persisted in believing that the first, third or fourth grooves were occasionally the origin of branchial cysts and fistulas. Kostaniecki and Milecki believed that Virchow's case in 1865 (35) originated from the first because of certain coexisting anomalies

of the ear. In 1894 Kostaniecki and Mielecki became dogmatic in their assertions that all median cervical fistulae were branchial in origin and not derived from the thyroglossal duct. They further stated that the outer openings of lateral fistulae of the neck could be found anywhere in the cervical region and that the external opening was of no import as regards to the origin of the fistula (19), (4). They contended that the outer opening of cervical fistulae developed from inflammation and suppuration, and consequent rupture of the inflammatory mass, might be at any height or position, depending on the dpeth or extension of the suppurating cyst or fistula. (19).

Again to the contrary Wenglowski in 1912 (36) stated that after studying a series of cases, one becomes cognizant of the fact that the external openings point in a definite place as a rule. That is, they occured along a line extending from the angle of the mandible to the middle of the sternum, along the medial border of the sternocleidomastoid muscle. This contention persists to the present day as pointed out by Christopher (12) (13) and Chemin (3). Wenglowski commented further on the statements of Kostaniecki and Milecki, that if suppuration and inflammation were factors in determining the external orifice, it would be

logical to expect it to point anywhere and not follow a definite course as we know it does. That is they could open lateral to the sternocleidomastoid muscle, in to it or even into the pharynx and esophagus. Wenglowski contended that the branchial apparatus does not spread itself into the viscera of the neck but belongs to the head (19).

Thus after reviewing the literature concerning branchial cysts and fistulae, we come to the point where a summary of the theories of origin of these conditions is in order.

1-The branchial clefts are responsible for all lateral cysts and fistulae,

(a) A fistulous opening or rest may persist from any of the four clefts advocated by (von Heusinger, Bland-Sutton, von Cusset and His).

(b) Only the second cleft can be at fault (Rabl).

(c) The cervical sinus is responsible for the external aspect of the anomaly as advocated by (von Ascherson, Kostaniecki, Milecki and His).

Then in 1912 Wenglowski came forth with the concept that the thymic stalk and not the branchial clefts were responsible for lateral cervical fistulae and cysts. Herbert Meyer in 1932 (27) entered Wenglowski's work

in the American literature and stated, that to explain the lateral cervical cysts and fistulae by the branchial theory is so artifical and shows so many erroneous conclusions that it seems we all ought to agree with Wenglowski and once and for all discard the branchial theory as the etiological factor. He went on to say Wenglowski's work proved that all cysts and fistulae above the hyoid bone might come from the branchial apparatus, and that everything below this level must come from other sources. Quoting Meyer's presentation of Wenglowski's work: "Anatomically, it seems impossible to think of the possible persistence of a cleft beyond the second embryological month. A cyst resulting from it is possible during the first or second embryological month, but not easy to imagine in adult life. The branchial structures obliterate so thoroughly that a complete fistula is not easy to imagine with the breaking through of a cleft to the outside. It is possible to imagine epithelial rests, but a complete fistula from a cleft is difficult to understand after the second embryological month. After that, in adult life, clefts cannot exist as the surrounding tissues continuously grow closer and closer together. The thymopharyngeal duct, however, will explain all the clinical findings. its course is exactly that of the findings in a patient.

Around the arch of the duct are the sinus rests. above the arch the duct forms a part of the third pharyngeal pouch. Below the arch it forms a part of the actual thymus duct. Microscopically, the fistulae and cysts coincide with the thymus anlage findings. One finds areas with stratified squamous epithelium mixed with ciliated epithelium, and in the walls encysted striated muscle fibers, cartilage and mueus glands. Lymphoid tissue is also present in large amounts which is never found in the midline ducts or cysts, but is found in thymus tissue and thymus ducts. It makes no difference where a thymus-duct cyst breaks through to the outside--either near the pharynx or near the sternum along the sternocleidomastoid muscle---the course of the thymus duct always remains the same. Lateral cysts and fistulae are rich in lymphoid tissue. This shows their close relation to the entoderm and to the thymus."

Wenglowski further stated that the stratified squamous lined cysts and fistulous tracts must undoubtely be of cervical sinus origin, while the anomalies that are entodermal are of thymic origin, and he felt that the dense lymphoid tissue with sometimes definite lymphoid follicles in the walls of the cysts and fistulae was strong evidence of the thymic duct origin (19).

Commenting on the thymic duct origin, Hyndman and Light

(19) stated that in view of the fact that cysts lined wholly by prickle cell stratified epithelium and therefore undoubtedly of ectodermal in origin, are commonly found exhibiting the dense band of lymphoid tissue, the latter could not be used to bear witness of thymic origin. Hyndman and Light in 1929 had studied the subject further and concluded that branchial cleft anomalies results from a failure of absorption of ectodermal and entodermal epithelium that is buried during the growth and fusion of the branchial arches in early life. This concept is the one that is accepted by most embryologists, surgeons and pathologists of the present date. There are however some men to this day who still insist that Wenglowski's thymic duct theory is the sole explanation of branchial cysts and fistulae. Lahey and Nelson in 1941 still maintain Wenglowski's theory should be accepted by all. (24)

EMBRYOLOGICAL HISTORY OF BRANCHIAL CYSTS & FISTULAE

The first accurate embryological description of the branchial apparatus in vertebrates was given by von Rathke in 1825. He described the arches and clefts in the three weeks' embryo of a pig and compared them with the branchial apparatus of the shark (19). In 1827, von Baer described four clefts in the human emb

ryo. (27) In 1832, Von Ascherson for the first time discussed the origin of fistulas of the neck from the branchial apparatus, while in 1877, von Cusset made a compilation of various pathologic conditions of the neck independent of the branchial apparatus. He claimed that in man the first arch develops on the fifteenth day and the fourth during the first half of the second month, and that all arches arise in the region of the base of the skull, but because of the rapid growith of the first, the lower four are crowded caudally. He further stated that the rims or borders of all four grooves or furrows are definitely delineated at first but soon fuse, and fuse on the outside first because they more rapidly here: and if the epithelium does not disappear before this fusion, many pathologic conditions would result. (19). In 1881, His, studying exclusively human embryos and applying the reconstruction method, changed considerably the study of embryology. He found that the endodermal and ectodermal layers come in close apposition with at least only a very small amount of mesoderm or partition between the pharynx and the outside in the region of the clefts. It is not risking too much, he stated, to suppose that this membrane might tear through and cause pathological conditions in the neck. (19).

His in 1881 stated that as one goes caudalward, the distance between the anterior ends of the arches becomes greater so that at one time a triangular area is formed with its apex upward and was termed by His the mesobranchial field. During further growth, the arches telescope themselves from above down so that looking from the outside, the fourth is covered by the third and the third by the second. In this way, the third and fourth become encased in a cavity in the wall of the neck by the downgrowth of the second arch. This cavity His called the precervical sinus. (19). Von Mall in 1887 agreed with His and Born. He found that in birds and higher vertebrates no breaking through of the branchial clefts took place, and that these clefts are always separated by a membrane composed of two epithelieal layers. In view of the fact that these membranes are thin and delicate it is possible, he stated, that they could rupture in the living embryo and thus give rise to many anomalies (19).

In 1888, Piersol, came forth with his study of rabbit embryos. He came to the conclusion that the inner pharyngeal pouches are developed earlier and mature later than the outer arches and clefts. In mammals, the rule is, according to him, that the branchial clefts and pharyngeal pouches are separated by limiting mem-

branes and that a tear can take place only in the second. In the first three pharyngeal pouches two parts can be distinguished, the ventral horn and the posterior extremity, while, in the fourth, the parts cannot be differentiated. Out of the ventral horns, with the exception of the first, the tubular epithelial formations grow (19).

Foll, von Liessner, Kostchenke, and Zimmerman took a stand opposite to that of His. In 1884, Forr stated that after studying a 6 mm human embryo that the branchial clefts must be considered as continuous structures, even though delicate limiting membranes are interposed (19). In 1889 von Liessner studying chick and sheep embryos concluded that patency of the branchial clefts does exist in all animals with certainty. Hammer in 1889 stated that in the 5 mm embryo, the pharyngeal pouches are well developed and the second is perforated bilaterally at its base (19). In 1912 Wenglowski published the monograph in which he gave the findings of the detailed work on human embryos, beginning with two millimeters and ending with forty nine millimeters, undoubtedly the most thorough work that was done on the subject up to that time. Therefore the author of this thesis feels that Wenglowski's embryology of the branchial apparatus should be pre-

sented and followed by Arey's embryology of the branchial apparatus, since the latter's presentation is the one that is generally accepted at the present time.

WENGLOWSK'S EMBRYOLOGY OF THE BRANCHIAL APPARATUS

During the second week of embryonic life, certain changes take place in the fetal foregut. From the interior lateral walls of the entoderm, five outpouchings or diverticula appear. These are called pharyngeal pouches. Simaltaneously the external ectoderm becomes indented over the corresponding pharyngeal pouches. These are the branchial or pharyngeal grooves. As the small grooves approach the pouches, the mesoderm is pushed aside so that for a time ectoderm and entoderm come into contact. The contacting areas are the closing membranes which in gill bearing animals disappear forming the gill clefts, opening from the pharynx to the exterior. Such perforation of course does not normally occur in birds and mammals. The grooves and pouches thus formed separate a series of rounded bars of mesoderm which has been pushed aside by them. These are termed branchial or visceral arches, of which there are six, all meeting ventrally in the midline of the neck. In each arch there is developed a cartilaginous bar consisting of a right and left half,

and in each bar also is the anlage of the primitive aortic arches. The first bar is called the mandibular arch, and from it are developed the lower lip, the mandible, the muscles of mastication and the anterior part of the tongue. Its cartilaginous bar forms the incus, malleus, and a portion of the mandible. The second arch is the hyoid arch. It gives origin to the structures of the upper part of the neck. From its cartilaginous are developed the styloid process, stylohyoid ligament, and the lesser cornu of the hyoid bone. The first two arches grow more rapidly than the remainder so that the latter four become telescoped, the first two arches leaving a deep depression, the cervical sinus. The ventral ends of the second along with those of the third arch assist in the formation of the body of the hyoid bone and the posterior part of the tongue. The third arch assists in the formation of the sides of the neck and its cartilage gives origin to the greater cornu of the hyoid. The fourth and fifth arches also assist in forming the sides of the neck, while the ventral portions of their cartilages unite to form the thyroid cartilage. The sixth arch gives origin to the cricoid, arytenoid and tracheal cartilage. From the first external indentation, the first branchial groove, arise portions of the auricle and external acou-

stic meatus, while of the second, third and fourth grooves, no traces persist. From the first internal outpouching, the first branchial pouch, are formed the auditory tube and tympanic cavity, the thympanic membrane arising directly from the closing membrane between the first and second arch. In the second pouch lies the anlage of the tonsil. Wenglowski demonstrated that from the third pouch on either side, a small duct, the thymic duct, descends into the mediastinum to from the thymus. This duct passes in the general direction of the sternocleidomastoid muscle laterally to the thyroid, and is lined with squamous epithelium surrounded by lymphocytes. From the fourth pharyngeal pouch a comparatively short tube passes on either side in a somewhat medial direction to form the lateral lobes of the thyroid. The parathyroids likewise originate in this pouch. The fifth pouch give origin to the ultimo branchial bodies, which although enveloped by thyroid tissue leave no identity in the human adult. (4).

AREY'S EMBRYOLOGY OF THE BRANCHIAL APPARATUS (3)

During the last half of the third week of intrauterine life the human embryo shows more clearly than at any other period a series of five parallel bars or visceral arches (branchial arches) obliquely placed on

each side of the cephalic portion in the region destined to become the upper part of the neck. These arches are separated from each other on the external surface by grooves termed visceral furrows (branchial clefts) and on the external surface by evaginations called pharyngeal pouches. Normally the intervening "closing membrane" between each groove and pouch does not rupture, so there is no communication between the visceral furrows and the pharyngeal pouches. From the first arch are developed the upper and lower jaw, the malleus and incus; from the second, the lesser cornu of the hyoid, the styloid process and part of the stapes; from the third, the body and greater cornu of the hyoid; from the fourth, the cuneiform cartilages and most of the thyroid cartilage; and from the fifth, part of the thyroid cartilage and the corniculate, arytenoid and cricoid cartilages. The first visceral branchial furrow or hyomandibular cleft, obliterates except at its dorsal part which becomes the external auditory meatus. The remaining furrows disappear. Internally, the first pharyngeal pouch gives rise to the eustachian tube and the tympanic cavity; the second forms no permanent organ unless a part is retained as the tonsillar and supratonsillar sinus; the third by entodermal outgrowth forms the thymus and the inferior

parathyroids; and the fourth the superior parathyroids. The tympanic membrane represents the dividing membrane between the first pharyngeal pouch and the first branchial cleft. Normally the arches become joined together anteriorly, that is on the ventral surface of the embryo. The first and second arches grow more rapidly than the others and overlap them forming the external groove called the cervical sinus or the precervical sinus of His. The thymus gland develops from two stalk-like d iverticula which originate during the sixth week from the third and fourth pharyngeal (branchial pouches). These diverticula grow caudad in the region destined to become the neck and fuse distally to become the thymic gland. During the few days in which these diverticula are patent, they are sometimes referred to as the embryonic thymic duct. Normally they are obliterated by the seventh week.

CLINICAL FEATURES OF LATERAL BRANCHIAL CYSTS & FISTULAE

A distinction is frequently made between branchial cysts and fistulas and each spoken as a distinct entity. It appears, however, that they are all simply a sinus tract which forms a cyst when the drainage is inadequate or obliterated. According to E. Andrews (1), there are five types of branchial cysts or fistulas;

1-Complete branchial fistula with internal and external opening.

2-Incomplete external fistula with no internal opening.

3-Incomplete internal fistula, with no external opening.

4-Branchial cysts with no internal or external opening.

5-Branchial dermoids.

The clinical picture of branchial fistula is that of a sinus which periodically drains a mucoid or thin milky material which may be associated with a cyst, depending upon adequacy of drainage. In children the fistula is also not prone to give any serious trouble in the average c ase except that the discharge is annoying to both parent and patient and the fistulous tract often stops drainage for a time with the production of a cyst which again periodically empties itself (4).

Not infrequently, however, repeated secondary infections do take place with an occasional serious cellulitis of the neck. Also some very aggravating conditions have been observed: a case reported by Louis Carp in 1926 (4) is that of a boy five years of age, who with a branchial fistula gave a history of a non-productive cough of two years duration which was unsuccessfully treated by tonsillectomy and medical measures.

At exploration an incomplete fistulous tract was found adherent to the vagus nerve. Excision of the tract brought about relief of all symptoms due to vagal irritation.

The fistulous opening may be situated at any level of the neck, but is constantly and without much variation just anterior to the sternocleidomastoid muscle. There is sometimes an area of pigmentation about the opening and occasionally a thickened bit of tissue may be felt which contains a bit of cartilage and is called the cervical auricle. The fistulous tract almost invariably runs upward and backward beneath the anterior portion of the sternocleidomastoid muscle. It passes over the carotid sheath to the midportion of the posterior belly of the digastric muscle whence it arches medially beneath the stylopharyngeus muscle to the tonsillar fossa. By tugging at the skin over the opening one can often feel the cord-like structure extending under the sternomastoid muscle (4).

The external opening may appear years after birth and is generally just anterior to the sternomastoid muscle at any level but most commonly in the lower third of the neck. A purulent discharge may occur externally or internally, and may vary in amount at different times. These sinuses can act as foci of infection and be

a detriment to the general health of the individual. When the cysts are large they may become objectionable because of their pressure on adjacent structues or because of their appearance. They may be the source of carcinoma (12). However Lahey and Nelson state that there has never been a case reported which was the source of carcinoma (24). Histologically the lining of the cysts and fistulas may be derivaties of ectoderm or entoderm, but both embryonic layers are occasionally thought to be present in the same cysts and fistula. In the latter event we must believe that an embryonic perforation existed between the pharyngeal pouch and the branchial cleft (19). Walls of the cyst most often is found to have a well organized stratified squamous epithelium resting on a relatively thick band of lymphoid tissue, while beyond this is connective tissue which may contain varying amounts of smooth and striated muscle which is usually infiltrated with elements of chronic inflammation, round cells and mononuclears. Occasionally columnar with ciliated cells are present. When columnar epithelium is present in the cyst there will be found thick mucoid transparent content. In the stratified squamous cyst the contents will be opaque, watery milky or cheese like. Though there are no reports on which to base an assertion of

a hereditary tendency to branchial cysts, there have been several instances of such a tendency to fistulas. The following are reported in Virchow's archives in 1861: mother and eight childre: mother and son in three instances: mother and daughter: two sisters: a brother and a maternal grandmother: aunts in two Instances(35). Hyndman reports: a mother and three of her five children presented fistulas, but there were no similar defect in the other members of her family. Hyndman makes the statement that branchial fistulas may be familial and hereditary, and seem to be in herited only from the mother.

SYMTOMS OF BRANCHIAL CYSTS AND FISTULAS OF THE NECK

Clinically a branchial cyst usually manifests itself simply as a uniform, painless, nontranslucent, semifixed fluctuant tumor which is never tender except when secondarily infected. In children and even in adults its presence does not cause them any particular discomfort. There have, however been reports of serious trouble caused by pressure on neighboring structures, particularly difficulty in breathing or swallowing. Thomson of England, reported a branchial cyst which produced hoarseness and was accompanied by a fixed vocal cord. When the swelling was at its height, there

was dysphagia and cough with complete misery of the patient. At exploration, the cyst was found attatched to the larynx and passed to the lateral wall of the pharynx. The symptoms completely sudsided after excision of the cyst (4). A branchial cyst as stated previously has a uniformly characteristic position along the anterior border of the sternocleidomastoid muscle and extends under it to the deeper structures of the neck. The size depends upon the amount of drainage or secretion present and may vary from a centimeter in diameter to one almost filling the entire side of the neck. The average size however is about that of a walnut. A fistula is complained of by the patient because of the continual discharge on the surface of the neck. A drop of watery pus is practically always present at the orifice of the fistula. Cases have been reported in the literature in which the fistula was of such size, patency and length that bread crumbs and other particles of food found their way down from the mouth out on the surface of the skin at the neck.

## AGE INCIDENCE OF BRANCHIAL CYSTS AND FISTULAE OF THE NECK

In 1929 Hyndman and Light (19) reviewed a series of ninety cases of cysts and fistulas. In their series the average age of the onset was seventeen years and

the condition was noticed at birth in only twenty-one percent of the cases, while eleven cases occured before the age of eleven years, which is the age limit of admission of the four cases reported from 1912 to 1921 in the Childrens' hospital of California (4). Shedden reported in 1931 the following data: (31).

| Age | Cases | Percentage |
|---|---|---|
| 1-10 | 4 | 8% |
| 11-20 | 19 | 37% |
| 21-30 | 15 | 29% |
| 31-40 | 6 | 11% |
| 41-50 | 4 | 8% |
| 51-60 | 12 | 2% |
| 61-70 | 2 | 4% |

## DIAGNOSIS AND DIFFERENTIAL DIAGNOSIS OF BRANCHIAL CYSTS AND FISTULAS

Shedden gives the following: fistulae are present at birth, situated at the lower third of the neck, may have ciliated epithelium and exudes clear mucus. While cysts appear in early adult life, have stratified squamous epithelium and not cliliated or columnar epithelium and contains opaque fluid. All fluctuant cystic swellings of the cervical region may be regarded as possible developmental anomalies. When they are inflamed and filled with pus, their cystic character may be difficult to recognize. The abscess may rupture internally and may burrow along the fascial planes of the

neck. The embryonic cysts may be distinguished from sebaceous cysts in that the latter are attached to the skin and are superficial to the muscles. Differential diagnosis must be made from inflammatory and tuberculous adenitis, hygroma, hemangioma and dermoids. Inflammatory adenitis is often bilateral and numerous glands are involved. They are tender and the offending focus usually can be found in sore throats, tonsillitis and teeth infections. Tuberculous glands may be more rapidly confused with cyst and fistula, but they are firm and tender and other glands are usually involved and often matted together in a group (4). Wangensteen (38) stated that 2 patients had come under his observation in which cervical cysts guise of other pathologic lesions over a long period of time. One patient was temporized for over twenty years under the impression he had tuberculous adenitis or a "cold abscess" from a tuberculous spondylitis which gave few physical signs. The other patient had been given a diagnosis of tuberculous adenitis and repeated aspirations had been practised when the "abscess threatened to breakdown". The diagnosis in both cases was made by Wangensteen to be branchial cysts by x-ray with injection of opaque medium. The discharge from the fistula in broken down glands in the neck is purulent while in branchial fis-

tulas the discharge is clear, mucoid or milky and may contain cholesterol crystals. As Shedden (31) states, a much neglected but valuable peice of evidence in the recognition of branchial cysts that has been significantly emphasized by Hamilton Bailey is the presence of cholesterol crystals on microscopic examination of the apsirated content. The high lipoid content frequently imparts a yellowish shimmer to the fluid when it is placed in an open basin. Though not invariably present these findings are practically conclusive for the existence of a branchial cyst. As for the differentiation of branchial cysts from cystic lipoma, the latter is translucent, occurs most frequently in the supraclavicular area, is often lobulated, and may grow to enormous proportions extending up to the face in front of the ear. Hemangiomata decrease in size on pressure and have the typical discoloration.

The diagnosis of fistulas may be facilitated by roentgenography after the injection of substances opaque to the x-ray (such as lipiodol), diluted half and half in liquid paraffin, with closure by purse-string suture after injection (4), or by the external injection of bitter substances to see if they may be tasted, or methylene blue to see if it appears in the throat. Traction on the skin may demonstrate the cord-like prol-

ongation of the fistula. The rising of the fistula with deglutition is said to signify that the fistula is complete. Cyts may be aspirated and injected with substances opaque to the x-ray, but they should be reaspirated afterward. If there is a swelling at the base of the tongue it shold be remembered that a cyst is thin-walled and light colored, whereas aberrant thyroid tissue is solid and dark red.

Visualization of the tract may be facilitated by a simple emthod described by McNeill and Love (4) and the procedure in my opinion seems to be of great value and will be described under treatment.

## TREATMENT OF BRANCHIAL CYSTS AND FISTULAE OF THE NECK

Chemicals, arsenicals and copper "flakes" perhaps were first used as powerful sclerosing salves in the treatment of superfical fistulae and even hemorrhoids, a method continued by quacks up to the present time. Dzondi wrote in 1829 (10) concerning a woman of twenty eight years of age whom he had seen in 1821 with a fistula in the neck one inch to the left of the mid-line. He states that after various caustics had been tried to no purpose, the fistula was at last healed by injecting 'liquor nitrici hydrargyri'. However he stated that later on the fistula reoccurred an the patient died in

1827 with the fistula still present. New inpetus to the sclerosing plan of treatment, has been given by Cutler and Zollinger who have apparently cured three cervical fistulas by in jection, one by Zenker's solution and two by Carnoy's solution (absolute alcohol, 6cc; chloroform, 3cc; glacial acetic acid lcc; and ferric chloride 1 gram) (10).

McNeill and Love gives the following method of treatment. A horse hair or dermal purse string suture is placed about the fistulous opening and drawn taut to shut off secretion. In a few days the tract becomes distended and identification is comparitively easy. The injection of methyline blue as ordinarily carried out, is usually a messy job. As the tract is opened somewhere in the course of dissection, the dye spills, stains all the surrounding tissue and all identification is lost. McNeill and Love, however, found methyline blue to be a distict aid when used in the following manner. The end of an ordinary hypodermic needle is broken and rounded off, the caliber of the needle depending on the size of the fistulous opening. The direction of the tract is determined with a probe and the methyl blue then injected with the use of the blunted needle. Too great a pressure may rupture the tract and defeat the purpose of the injection.

At the same time the fingers of the left hand gently massages the neck to and fro in the direction of the tract. Thus the epithelium in even the minutest diameter of the tubule may be reached and stained. Using the same syringe and needle we then gently but thoroughly wash out the methyl blue with water or saline, alternately massaging outwardly until the return is only faintly stained. In this manner the epithelial lining remains stained and if the tract is inadvertently opened during the dissection, no soiling of the neighboring structures will take place. A small probe is again inserted and the incision is made thru the skin and platysma muscle along the anterior border of the sternomastoid and surrounding the fistulous opening. (The above incision is no longer used and is replaced by the incision oblique and parallel to the skin creases of the neck) (12). The platysma is reflected laterally on both sides, and the sinus is dissected to the pharynx in its entirety. The stump may be dealt with by several methods as recently described by Meyer (27). By the method of von Hackerer a probe is passed through the lumen of the duct, into the pharynx, and with suture the stump is attached to the probe and inverted into the mouth cavity and there tied and cut. By the method of Koenig a probe is forced into the pharynx

a short distance from the duct which is then sutured and pulled with the probe into the pharynx, thus re-implanting it. The method of von Hackerer appears most desirable for several reasons: there is not the possibility of hemorrhage from the passage of the probe, and there is not the dangerof opening fresh avenues of infection from the mouth and lastly according to McNeill and Love it does away with the entire tract. McNeill and Love and Baumgartner (4) state that the dissection of the stump may be carried to the pharynx by simply having an assistant place a finger in the tonsillar fossa, and having the assistant push outward, the stump is easier to dissect. When the dissection reaches the assistant's finger the stump is severed and touched with tincture of 10% iodine. The wound is close in the usual manner.

Christopher (12) states the following concerning treatment of lateral cysts and fistulas of the neck. "Radical extirpation of the cyst or fistula is regarded by most surgeons as the only method of treatment which offers a chance of certain cure. This, at times may be a formidable operation and should be undertaken only in a hospital operating room. The incision should oblique and parallel to the skin creases in the lateral cysts and fistulas. The pharyngeal end of a complete

fistula may be inverted into the pharynx and the fistulous tract closed by suture. Where access to the cyst is difficult the sternomastoid muscle may be divided. Preliminary aspiration is thought by some to facilitate excision of a cyst. In some of the cysts which are lined by mucous membrane the cyst wall may be very friable and difficult to remove. If such wounds are packed with gauze the cyst wall may come out when the packing is removed".

Ladd and Gross (25) give this word of caution, and rules to follow. If their is any question of the cyst containing solid tissue it should be opened before detaching it from its bed for it may be an ectopic thyroid. If found to contain thyroid tissue, leave it in its abnormal position, close the wound: otherwise mxyedema will result. For cases have been reported that since this ectopic thyroid is usually the only thyroid tissue present, its removal will cause mxyedema within a very short period of time. Hicken and Popma in 1938 (21) also report that removal of ectopic thyroid tissue for branchial cysts will result in myxedema. Thus it is necessary to include ectopic thyroid tissue in the differential diagnosis of branchial cysts of the neck.

## HISTORICAL BACKGROUND OF THYROGLOSSAL DUCT CYSTS AND FISTULAE

The historical background of the thyroglossal duct cysts and fistulae in comparison to that of the branchial apparatus anomalies has received very little consideration as far as the available literature on the subject is concerned. Verneuil in 1853 (2) described cysts in the thyro-hyoid region, which he ascribed to thyro-hyoid bursae, of which he found several. Others are inclined to think that his cases were true thyroglossal cysts. His (2) a few years later, whose researches into the embryology of the thyroid gland and its dvelopment are well known, was the one who gave the name "thyroglossal duct of His" to the tube which developed in consequence of the descent of the thyroid gland. Other pioneer embryologists who investigated the development of the thyroid gland besides His during the middle of the eighteenth century were von Rathke and Kollicker (37). All these men agreed that in consequence of the descent of the thyroid gland a duct was formed, but which soon was obliterated as the thyroid gland developed further. In the later part of the eighteenth cenury there was considerable confusion as to the matter of fistulae and cysts in the mid-line of the front of the neck. His (19) had pointed out in 1891 c-

ases in which the persistence of the thyroglossal tract was definitely the cause of many midline neck fistulae and cysts. Durham (15) in 1894 reviewed the literature printed up to that time and pointed out that Lahneloongue and Archard were inclined to regard all median cervical fistulae as branchial in origin. He pointed out that at that time von Kostanieck and Mielecki and Kanthack had been most dogmatic in their assertions that all median cervical fistulae were branchial in origin and not to the persistence of the thyroglossal tract. The latter three mentioned men further stated the "sinus precervicalis of His", a cavity formed by the overlapping of the hyoid arch in front of the third and fourth arches, was the sole cause of median cervical cysts and fistulae (19). They even went further in their assertions, stating that the only difference there was between median and lateral fistulae was the median position of the aperature on the neck of the median form. Durham then stated that the diagnosis of sinus precervicalis fistulae or cysts could be made only when squamous epithelium occured about the aperature of the fistula, since that structure as he stated was derived from the epidermis of the branchial system. Durham then asserted that Kostaniecki and Mielecki had tabulated median branchial cysts and fistulae but which

were in reality of thyroglossal in origin. Durham however closed the contraversy, by stating that the persistence of the sinus precervicalis could be one other possible cause of median cervical fistulae.(36).

The other pioneer embryologists who studied the development of the thyroid gland, and who pointed out that the failure of obliteration of the "thyroglossal duct of His", caused median cervical cysts and fistulae, were von Rathke and Kolliker. The etiological factors pointed out by the above mentioned men, concerning median cervical cysts and fistulae has not changed even to the present time.

## EMBRYOLOGY OF THE THYROID GLAND

The pioneer embryologists who studied the embryological development of the thyroid gland and who established the fundamental facts concerning this gland which are well known even to the present day, were His von Rathke, and Kolliker. The first report devoted entirely to a study of the embryology of the thyroid, that of Tourneux and Verdun in 1897 (37) was based on a series of eight human embryos and five fetuses. They concluded that the thyroid gland arose from one medial and two lateral bodies. The first appearance of the median thyroid was indicated as appearing at the 3 mm

stage. This primordium then underwent a series of changes becomingly successively enlarged, lobulated bilobed and detached from the pharynx, transformed into vascularised anastomosing cords and located in the 16 mm embryo. Its typical position draped across the anterior portion of the pharynx. The lateral thyroids, arising from the anterior walls of the fourth "entodermal pouch" underwent, simaltaneously, detached from the pharynx and the pharyngeal cavity, after which they fused with the median thyroid. In 1933 Weller and Norris, after thoroughly investigating the subject, agreed with the work of Tourneux and Verdun. Weller and Norris concluded that the fourth pharyngeal pouches called ultimobranchial bodies by many embryologists, contributes significantly to the definite thyroid gland (3). Kingsbury states that the ultimobranchial bodies donot form the lateral thyroids, and that these bodies completely degenerate and disappear (3). Because of the thorough work done by Weller concerning the developmental stages of the thyroid gland his investigations will be presented in this thesis.

## WELLER'S DESCRIPTION OF THE DEVELOPMENTAL STAGES OF THE THYROID GLAND

The first evidence of the presence of the median

thyroid component is a well defined fold in the ventral wall of the pharynx. This occurs before the second pair of somites has been definitely separated off and consists in a proliferation, rather than primarily a differentiation of pharyngeal cells. This fact seems not to have been noted or at least not to have been stressed by previous observers as indicating the first appearance of the median thyroid primordium, but it is of considerable importance because throughout subsequent development of the thyroid gland evidences of cellular proliferation always precedes cellular differentiation. As growth continues the medial thyroid undergoes changes in its external form as the result of which it assumes a spherical shape. It is located on the ventral aspect of the pharynx, midway between the primordia of the tonsils. Its walls and lumen, continuous with those of the pharynx, are composed of two types of cells. With further growth of the embryo, the distance between the median thyroid and ventral wall of the pharynx increases. Simaltaneously, there occurs a change in the shape of the median thyroid component so that it assumes an inverted Y appearance. By the time the embryo has become 14 mm in length, the median thyroid assumes the U shape which is of so much importance in the formation of the definitive thyroid

gland. It is still impossible to designate the lateral portions as true lobes, nor is the isthmus composed of the tissue which it will contain in the adult. The structure, sheet like in form, is draped across the pharynx and curled around the external carotid arteries.

The lateral thyroid components are evidenced by proliferation of certain cells of the pharyngeal endoderm. In the 20 somite embryo it is possible to identify with certainty the lateral thyroid components. IN this embryo, the histological changes in this region are indicative of only beginning proliferation of the lateral thyroid cells. The latter are still identical with the cells of the pharynx, but in their formation have formed outpouchings of the pharyngeal wall. These pouches are the lateral thyroid primordia. At the outset, the chief difference between the three thyroid primordia is one only of location. This similarity is emphasized by the configuration of the floor of the pharynx in this region. Further, each of the components is hollow and its lumen is continuous with that of the pharynx. Just as this arrangement emphasizes the similarity between the thyroid primordia, so it emphasizes the individuality of the latter in contrast to the other primordia. The lateral thyroids become increasingly further removed from the pharyns so that by the

time the embryo reaches a length of 10 mm. the lateral thyroid primordia are shaped, in general, like a rounded long-necked flask. The three primordia grow in a longitudinal direction, their pedicles become severed and a layer of mesenchyme develops between them and the pharynx. Growth of the lateral thyroid cells proceeds however at a much slower rate, for even in embryos 16 mm. in length, the lateral thyroids are still attached to the pharynx. The fusion of the median and lateral thyroid components is as follows. The lateral thyroids move upward and backward a slight distance, still, however, maintaining its intimate relation to the internal carotid arteries. The lateral thyroid is approximated to the posterior surface of the medial thyroid, but the cells of each remain separated by a delicate layer of connective tissue. The 23 mm. embryo first exhibits the external form which will characterize its gross anatomical appearance throughout the remainder of fetal as well as post-fetal life. It consists of the usual lobes, located on either side of the pharynx, and connected by the narrow isthmus. Adjoining it on either side of the pharynx, posterolaterally, is the cervical portion of the thymus gland. Both parathyroid glands are located upon the posterior surface of the lobes of the thyroid gland.

Thus from direct embryological evidence, which dovetails perfectly with the findings in post-fetal life, it may be concluded that the thyroid gland of man assumes its adult architecture in the following manner. Three primordia, one median and two lateral after a series of progressive changes in growth and differentiation, fuse into a morphologically single structure.

AREY'S EMBRYOLOGY OF THE THYROID GLAND AND TRACT

The thyroid gland develops from an outpocketing from that portion of the floor of the pharynx whose location in later life is marked by the foramen cecum on the posterior portion of the roof of the tongue. This epithelial evagination is transformed into a stalked vesicle by the time the embryo has reached a length of 2.5 mm. This vesicle or diverticulum grow downward and backward from its point of origin as a tubular (thyroglossal duct) which bifurcates at its distal portion to form the thyroid lobes. During its transitory existence, the duct proper passes through or adjacent to an area which at a later time becomes the hyoid bone. Its distal end corresponds to the region which later becomes the pyramidal lobe of the thyroid. This epithelial lined duct usually atrophies during the

sixth week of fetal life; emnants of an epithelial cord persist for a short time. The parathyroid glands are epithelial bodies arising from the posterior wall of the third and fourth pharyngeal pouches.

## CLINICAL FEATURES OF THYROGLOSSAL CYSTS & FISTULAE

Hicken and Popma (21) state that thyroglossal cysts and fistulae arise from sequestrated epithelial rests which have been torn from the floor of the mouth by the descending thyroid anlage and deposited along the thyroglossal tract. Christopher (13) states that cysts and fistulas in the mid-line of the neck are remnants of the embryonic thyroglossal duct. Because they are intimately attached to the hyoid bone and the foramen cecum, these tumors always move upward on deglution or extension of the tongue (21). Callander (14) states that thyroglossal duct cysts usually persist between the foramen cecum and the hyoid bone, and that portion of the duct behind and below the hyoid bone sometimes develops into a sub-hyoid cyst which may rupture and become a thyroglossal sinus. Concerning the mechanism which brings about thyroglossal duct cysts & fistulae, Jarvis (22) states the following: Coincidentally with the development of the thyroglossal tract & as a consequence of its rapid growth, small segments of

the normal lining embrane of the pharynx about the primordium of the thyroid gland are torn loose and carried for varying distances into the depth. These segments have all the tissue elements of the mucosa and and of these the important structures are the epithelium, the glandular elements, and the lymphoid tissue. The complex relation of these cysts with the hyoid bone has been a matter of careful examination. The thyroglossal tract and the hyoid bone are developed before the muscles and fascia attached to the bone appear. The thyroid tissue and the associated tissue of the pharynx are thus in very intimate connection with the hyoid bone. As the muscles and fasciae appear and competitive growth begins, the weaker thyroid epithelium tends to atrophy, but the more vigorous mucosal epithelium advances, and may also grow into the periosteum and may erode the hyoid bone substance (22).

Bertwistle and Frazer (8) sums up the clinical features of this condition as follows: The thyroglossal remnants above the hyoid are few and, if present are usually either near the foramen or near the hyoid bone. Below the hyoid, the tract is present completely in many cases, and incompletely in others. The complete infra hyoid tract extends from the hyoid usually folded up. The incomplete tract has potentially and exactly

the same line as the complete ones: its tract is simply broken up and its parts remain in situ. Any one of these remnants may take on further development, especially perhaps if a condition of relative hypo thyroid exists. It is possible that some cells of the tract may be separated off and lodged below the level of the gland: if this is so, these cells may also be liable to take on further development. They also pointed out that certain embryologists 40-50 years ago, maintained that the median fistulae etc, found low down in the neck arose from remnants of the precervical sinus, a view strongly supported by Kanthack in this country. This view would not receive extensive support from embryologists today and may be passed by (8). Cysts lying superficially below and behind the foramen cecum may have possible origin from a space included between the halves of the developing hinder part of the tongue, and not necessarily be connected at all with the thyroglossal tract.

A case of thyroglossal cyst might be explained by a patent thyroglossal duct. If such a duct opening into the mouth at the foramen cecum becomes for some reason or another occluded, then a thyroglossal cyst might be expected to result. Bailey (5) stated that such a theory postulates a patent thyroglossal duct opening into the mouths of a certain percentage of normal ind-

ividuals for it would be unreasonable to suppose that it always becomes occluded. Furthermore this theory entirely fails to explain the parallel case of branchial cyst which in the vast majority of cases is quite unconnected with the pharynx (5). Bailey gives his own theory and is as follows: "Thyroglossal cysts and branchial cysts are not infrequently surrounded by lymphatic tissue. In common with the neighboring lymphatic system this from time to time becomes the seat of bacterial invasion. If this is so then the lining epithelium is liable to become involved and reacts to the infection in just the same manner as other epithelial surfaces (notably the nasal mucus membrane by pouring forth secretion. Some evidence in favor of this theory is as follows: 1-Four cases of thyroglossal cyst appearing in adolescent or adult life commenced with tonsillitis or pharyngitis for which the patient had received treatment. Several more gave a history of sore throats which may or may not be relevant. 2-A baby admitted with an infected foot developed a thyroglossal cyst which suppurated. 3-There is overwhelming proof both clinical and histological to show that thyroglossal cysts are frequently the seat of inflammation". (5).

Jarvis (22) states that pathologic conditions found in post-natal life that seem to have their origin

in vestigal rests of the thyroglossal tract and its inclusions are: 1-Heterotopic masses of thyroid tissue in the root of the tongue (lingual goiter), or above or below and rarely in front of the hyoid bone. 2-Median cysts in the tongue, under the tongue, and above about or below the hyoid bone. 3-Median fistulae.

A thyroglossal duct cyst is a retention cyst which arises in a patent portion of the vestigial thyroglossal tract and occurs anywhere in the midline along its pathway from the base of the tongue to the region between the hyoid bone and thyroid gland. Clinically the site of the cysts is more common below the level of the hyoid then above it. Frequently the cyst ruptures through the skin spontaneously or is incised and an intermittent draining channel which may end blindly in the tissues, that is, a sinus may result from the rupture, or the channel may run uniterruptedly to enter the mouth through the foramen cecum. That is a fistula. A knowledge of the origin and development of the thyroid gland signifies the understanding of the surgical treatment of the thyroglossal duct cysts and fistulae. In as much as the hyoid bone develops either simultaneously with, or later than, the descent of the thyroid gland, the thyroglossal duct may vary in its relation to the hyoid bone. The tract lined with epithelium prod-

uced by this descent normally disappears between the fifth and eighth weeks of fetal life, but occasionally fails to become obliterated. This failure of obliteration of either a portion of, or of the entire, thyroglossal duct subsequently may lead to the formation of a thyroglossal duct cyst, sinus and rarely a fistula.

LOCATION OF THYROGLOSSAL DUCT CYSTS AND FISTULAE

They may occur anywhere from the foramen cecum of the tongue to the body of the thyroid gland.

1-Beneath the foramen cecum.

2-In the floor of the mouth protruding beneath the chin.

3-Suprahyoid.

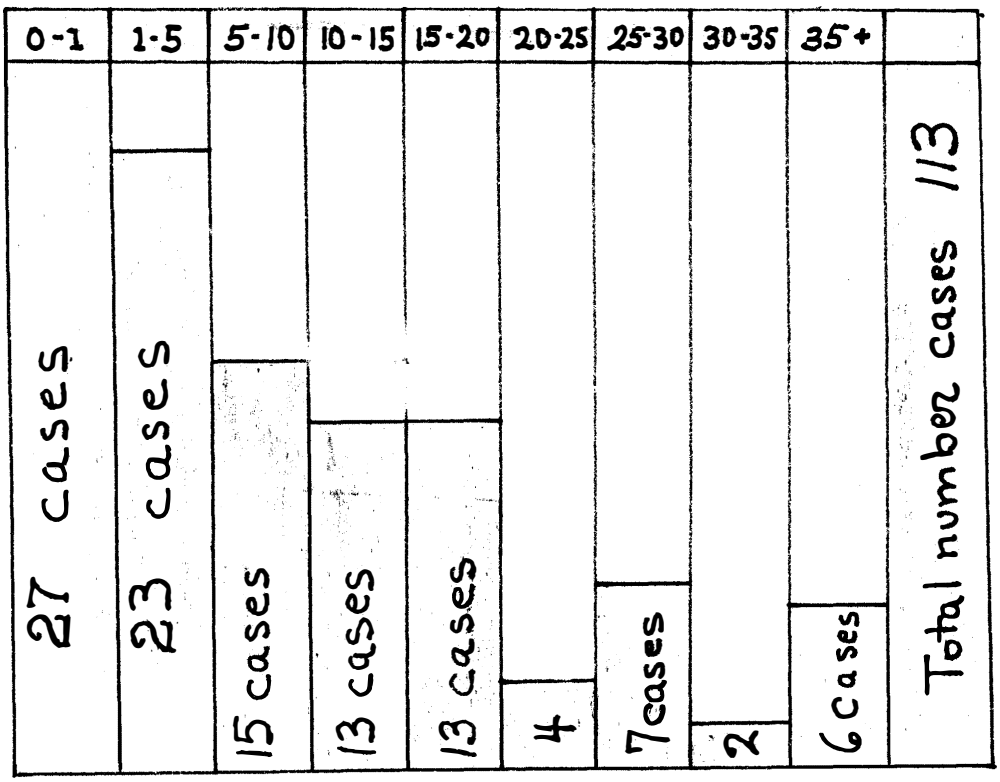
4-Subhyoid.

5-On the thyroid cartilage or membrane.

6-At the level of the coracoid body in the suprasternal notch (22).

It has been almost invariably taught that thyroglossal cysts and fistulae are located in the median line of the neck somewhere along the course of the thyroglossal tract. This need not be the case. One of Jarvis' cases extended to the left of the suprasternal notch, and in this respect Jarvis stated that both the opening of the fistula or cyst may be eccentrically placed, but the

AGE INCIDENCE OF THYROGLOSSAL DUCT CYSTS & FISTULAE




Age at which the thyroglossal cyst or fistula first appeared. Report of 113-cases by Bailey (7).

In the cases reported through out the literature this graph is closely met by all (22), (8), (5), & (29).

In contradistinction to branchial cysts which most frequently appear in early adult life, thyroglossal cysts are commonly met with in babies, and as the figure shows the majority of cases reviewed occurred in infants or the history of trouble dates back to very tender years. To recapitulate: thyroglossal cysts sometimes, and branchial cysts generally make their first appearance after the second decade of life. The cases reported at the University Hospital at the Nebraska School Of Medicine showed the same age incidence of the two conditions.

PATHOLOGY OF THYROGLOSSAL DUCT CYSTS AND FISTULAE

The two conditions may be lined by ciliated columnar or flattened epithelium. But may undergo alteration as the result of chronic inflammation. The walls may be composed of lymphoid tissue showing in some instances round cell infiltration (29). The walls may contain embryonic thyroid tissue. A purulent discharge may occur externally or internally and may vary in amount at different times. Christopher (12) states that they may be the source of carcinoma although Patterson and others state that no case of malignant disease has been reported. The tract may run in front of, in, or behind the hyoid bone.

## SYMPTOMS OF THYROGLOSSAL DUCT CYSTS & FISTULAE

Rarely does a thyroglossal cyst when situated in the neck give rise to symptoms. Symptoms may occur as the result of inflammation in the cyst, causing the skin discoloration, tenderness over the swelling and pain especially on swallowing. The skin may become red and adherent, and the cyst wall may eventually rupture with the formation of a fistula. Cysts of the upper end of the thyroglossal tract may interfere with the functions of speech, deglutition and respiration and cause anemia from recurrent hemorrhages. Wardill reported (29) a case in which the cyst was filled with pus which could be squeezed from the patent foramen cecum of the tongue. The excretion from a fistula is never copious (29). It may be watery, syrupy, mucus-like or purulent: and it may contain iodine.

## DIAGNOSIS & DIFFERENTIAL DIAGNOSIS OF THYROGLOSSAL DUCT CYSTS AND FISTULAE

Jones (23) states that one most remember the dogmatic teaching that all midline cervical cysts and fistulae are to be considered as coming from the thyroglossal duct until proven otherwise. Durham states that the diagnosis depends on the position strictly in

the midline; the presence of ciliated cylindrical epithelium which may be mor or less replaced by squamous epithelium as the upper portion of the tract is approached; permeability to injection or probes through the foramen cecum; the presence of paried hemina, the remains of the original bifurcation; the presence of thyroid gland follicles, or accessory glands intimately associated with the duct; and lastly the connection with the thyroid gland at the lower extremity.

Diagnosis of such cysts is usually not difficult and is made by the finding of a rather firm, cystic tumor in the midline of the neck near the hyoid bone or the thyroid cartilage. When this is palpated the duct which runs from the cyst to the hyoid bone may usually be felt. If the cyst is left alone, it gradually enlarges and often is drained surgically. In other cases infection occurs within the cyst and an abscess forms which also is often opened and rained. In either case a sinus remains which discharges the fluid secreted by the epithelium lining the tract. In many of the patients Sistrunk (33) had examined the fistulas had been present and persisted for periods varying from six months to twenty-nine years.

If a thyroglossal cyst comes under observation for the first time when complicated, its true nature is

easily over looked and a diagnosis of some inflammatory condition is made, notably an abscess 'en bouton de chemise' connected with a tuberculous pretracheal gland. Again in the midline a simple epidermoid cyst is a difficult but an unimportant differential diagnosis. In this connection it should be mentioned that a thyroglossal cyst quite frequently moves upwards on fully protruding the tongue, and this sign may serve to distinguish it from an epidermoid cyst. Lastly, a diagnosis of a thyroglossal cyst is never complete with out definitely determining the presence of a thyroid gland in its normal position, for an ectopic thyroid has probably many times been unwittingly removed. Not infrequently this thyroid is the only thyroid for as Spencer (5) states "the persistence of thyroid tissue in the course of the thyroglossal tract stands as an inverse relationship to the normal development of the normal thyroid gland".

The diagnosis of fistulas may be facilitated by roentgenography after the injection of substances opaque to the x-ray (such as lipiodol), diluted half and half in liquid paraffin, with closure by purse-string suture after injection, or by the external injection of bitter substances to see if they may be tasted, or methylene blue to see if it appears in the throat.

Sebaceous cysts are attached to the skin and are more superfical. Lipomas are softer and generally presents the "signe de peau d'orange" (12). Softened tuberculous lymph glands or inflammatory adenitis should not be confused. If there is a swelling at the base of the tongue it should be remembered that a cyst is thin walled and light colored, whereas aberrant thyroid tissue is solid and dark red. Cysts may be aspirated and injected with substances opaque to the x-ray, but they should be reaspirated afterward.

TREATMENT OF THYROGLOSSAL CYSTS AND FISTULAE

Of the congenital anomalies commonly encountered in the cervical region perhaps none have resisted surgical correction for a long time as have the cysts, sinuses and fistulae arising from a persistent thyroglossal duct. The reasons for this are not entirely clear. Since the nature of the dvelopmental effect offers little technical difficulty it seems incredible in retrospect that surgeons continued to employ with so little success inadequate measures until 1920 when Sistrunk advised excision of the entire thyroglossal tract and devised for this purpose a safe and thoroughly satisfactory operation. Sistrunk was not one of the first to preform a radical proceedure in these cases.

Schlange in 1893 and Durham 1894 (30) cognizant of the fact that unsuccessful attempts at cure were due to incompletness of the operative removal of the tract, divided and removed a segment of the hyoid bone and dissected the duct to the root of the tongue. Later Elliot and Spencer advocated similar procedures. However Sistrunk recognizing that it is not always possible to trace the upward extension of the tract, even in cases in which the sinus has been injected with methylene blue, devised a procedure that would not fail to completely remove the entire tract and advocated its employment as routine.

## THE SISTRUNK OPERATION OF THYROGLOSSAL DUCT CYSTS AND FISTULAE

Under general anesthesia, a transverse incision two to three inches in length is made at about the level of the hyoid bone. When a sinus is present, it may be injected with methylene blue to outline its course, and an elliptic transverse incision made to include the sinus. The skin, subcutaneous tissue, and platysma muscle are reflected. The cyst or sinus will usually be found lying on the thyro-hyoid membrane. It is dissected free from the surrounding tissues up to the hyoid bone. The relation of the tract to the hyoid

bone is variable, usually passing through or beneath the bone, but occasionally passing above it. For this reason to insure complete removal of the tract at this point and to facilitate exposure above this point the central portion of the hyoid bone is freed above and below, and the mid-portion, measuring about 1/4 inch in length, is removed with bone forceps. As a rule the dissection up to this point is carried out without great difficulty, but above this the tract is usually so small and friable that it is broken off easily and is difficult to remove. For this reason no attempt is made to isolate the tract, but the duct and tissues surrounding it for a distance of three or four mm. on all sides are cored out through themuscles of the tongue to the foramen cecum. This can be done without difficulty as Sistrunk has shown that the pathway of the duct corresponds to a line drawn at an angle of forty-five degrees backward and upward through the point of intersection of lines drawn horizontal and perpendicular to the center of the bone. This dissection can be facilitated by placing the index finger of the left hand in the patient's mouth and pushing the foramen cecum upward and forward. A portion of the hyoid bone, portions of the raphe of the mylohyoid muscles and portions of the geniohyoid and genioglossos muscles are

removed. In cases in which the duct is patent above the hyoid bone the foramen cedum is also removed, but in others in which the cyst seems to be confined entirely to the sub-hyoid region it is satisfactory to stop the dissection just at the foramen cecum without removing it and consequently without entering the mouth.

A series of 293 cases of thyroglossal duct cyst in which the Sistrunk operation was employed were reviewed. (30) 261 of these cases were satisfactorily traced to the possibility of recurrence. There were four recurrences in this series. The recurrences for the most part can be attributed to the presence of extensive scarring and infection as a result of previous surgery. The effectiveness of the Sistrunk operation stands out in contrast with many failures common to the less radical procedure.

Gessner (18) doesn't believe in the Sistrunk operation to the foramen cecum because he feels that the removal of the tube too near the foramen will permit the discharge of secretions into the mouth and so obviate the recurrence of the fistula. He however did-not give reference to any cases in which such a condition occured.

Gill (17), Payne (28) and a host of other men

to numerous to mention, have pointed out also that only radical surgery can prevent recurrences. At the same time these two men and a host of others state that cautery and sclerosing methods should be mentioned only to be condemned.

Bailey (7), Pemberton & Stalker (30) at Mayo's, Bertwistle & Frazer (8) in London, Shedden (31) and many others who have dealt with numerous cases of thyroglossal duct cysts and fistulae, all have reported great success with the Sistrunk operation.

## CONCLUSIONS

In spite of the great work done by Wenglowski on the branchial apparatus, and his theory that the thymic stalk is the primary cause of branchial cysts and fistulae, I am inclined to believe that the conditions are the result of a failure of absorption of the epithelium that is buried during the growth and fusion of the branchial arches in early embryonic life. The reasons for my excepting the branchial theory are; fistulae openings at the region of the auricle of the ear, & at the angle of the jaw cannot be explained by the thymic stalk theory, whereas the branchial theory can explain them together with other lateral fistulae and cysts of the neck; since there are five arches and only

one thymic stalk on either side of the embryo's neck region, the propertion shows that the changes for the branchial arches and clefts to form anomalies are five times greater than that of the thymic stalk; and finally since the presence of stratified squamous epithelium in such anomalies cannot be explained on the thymic stalk theory, it can be explained by the branchial theory. As for the treatment of branchial and thyroglossal cysts and fistulae, there is only one treatment and that is radical surgery. This fact speaks for itself after reviewing the literature which points out the excellent results produced by such a procedure. I believe that Sistrunk's operation for thyroglossal cysts and fistulae is an excellent one, and it should be done routinely for such conditions in order to insure good results. Sclerosing solutions & cautery in my estimation should never be attempted to cure any of the cysts and fistulae of the neck, because the changes of recurrence are too great.

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